



CASE REPORT

Solid Pseudopapillary Pancreatic Tumor: Added Value Contrast-enhanced Ultrasound in Diagnosis and Follow-up

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Solid pseudopapillary tumor (SPT) is a very rare neoplasm of the exocrine pancreas. SPT usually develops asymptotically and therefore radiological techniques are very important in the diagnosis. Frequently, ultrasound examination is the first technique that is used, and although it is not very specific, it may be completed with contrast-enhanced ultrasound, which increases diagnostic accuracy. Unlike other pancreatic neoplasms, SPT has a homogeneous filling in the arterial phase and is well defined by peripheral rim enhancement in the early dynamic phase. We report the clinical and imaging features of a case of SPT, as well as the surgical treatment and postoperative complications.

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Introduction

Solid pseudopapillary tumor (SPT) is a very rare neoplasm of the exocrine pancreas that mainly affects young women. If

diagnosed in time, it can benefit from curative surgery with excellent survival rates. The main diagnostic tools are radiological techniques. Ultrasound (US) examination is usually the first technique that is used and may suggest the diagnosis. Moreover, US is important in the postsurgical setting to evaluate the possible complications. Abdominal computed tomography (CT), magnetic resonance imaging (MRI) and endoscopic ultrasound-guided fine-needle aspiration (EUS-FNA) are more accurate diagnostic tools.

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In this paper, we describe the clinical and imaging features of a case of SPT, as well as the surgical treatment and postoperative complications.

Case report

A 20-year-old woman, with no remarkable medical history, was admitted because of a 1-month history of epigastric pain, nausea and vomiting. Her physical examination was unremarkable, except for epigastric tenderness. The only laboratory test abnormality was a slight increase in serum amylase at 148 U/L (normal value <100 U/L) and discrete augmentation of carbohydrate antigen 19-9 antigen at 32.69 IU/L (normal value <19 IU/L).

The US examination (Logiq 7; General Electric, Milwaukee, WI, USA) revealed a 92 mm × 58 mm × 43 mm, mixed solid and cystic mass in the pancreatic body and tail (Fig. 1A). The tumor was well-defined and had a close

contact with splenic and superior mesenteric veins. Color Doppler examination confirmed the permeability of these veins. Intravenous contrast-enhanced ultrasound (CEUS) in pulse-inversion mode at a mechanical index of 0.09, after 1.6 mL bolus injection of the contrast agent SonoVue (Bracco, Milano, Italy) showed a homogeneous vascular pattern of the solid part of the tumor, with a relatively slow washout (28–30 seconds) compared to the normal pancreatic parenchyma (Fig. 1B). The tumor was well-defined by an enhanced rim around the mass in the early phase. There were no signs of local extension or hepatic metastases in the portal phase. No other pathological findings were described at US examination.

EUS-FNA was non-contributive because of the lack of sufficient biopsy material. However, endoscopic ultrasound was able to eliminate local tumoral extension.

During laparoscopic surgery, a pancreatic tumor was identified extending from the body–tail pancreatic junction to the splenic hilum. Given the close contact of the tumor with the splenic vessels, their excision was necessary. The spleen vascularization was kept through the gastro-splenic ligament, permitting sparing of the spleen.

Histological examination revealed an SPT of the pancreas, with specific immunohistochemical profile (vimentin, neuron-specific enolase and CD10 positivity).

In the postsurgical survey, no signs of sepsis or hemodynamic instability were noted. After splenic vessel branches excision, a control US examination was made 7 days after surgery. This revealed a well-delimited liquid collection in the pancreatic area, which was interpreted as a postsurgical hematoma (Fig. 2A). The spleen volume was significantly increased, with a well-delimited hypo-echogenic area situated at the inferior pole. CEUS excluded an active retroperitoneal hemorrhage and the lack of contrast enhancement in the hypo-echogenic splenic area suggested a splenic infarction (Fig. 2B). The patient was asymptomatic and therefore conservative management was chosen. Given the favorable evolution, the patient was discharged. Evaluation at 6 weeks and 3 months showed resolution of the postsurgical hematoma and scarring of the spleen infarction lesion.

Discussion

SPT of the pancreas is a very rare neoplasm, with a low malignancy potential [1]. It represents 1–2% of the pancreatic neoplasms and is more frequent in young women [2]. This neoplasm rarely affects men, in which case it has the tendency to develop 10 years later than in women [3]. A recent review of the medical literature has described 718 well-documented cases of SPT between 1933 and 2003 [4].

This neoplasm was first described by Frantz in 1959 [5], and the term SPT was recently established by the World Health Organization working group for studying pancreatic tumors [6], after a period of many years during which this tumor was named in many different ways.

SPT usually develops asymptotically, but when symptomatic, the most frequent symptom is abdominal pain, followed by evidence of an upper abdominal mass at the physical examination. The first symptom may rarely be jaundice, if the tumor is situated in the pancreatic head. At

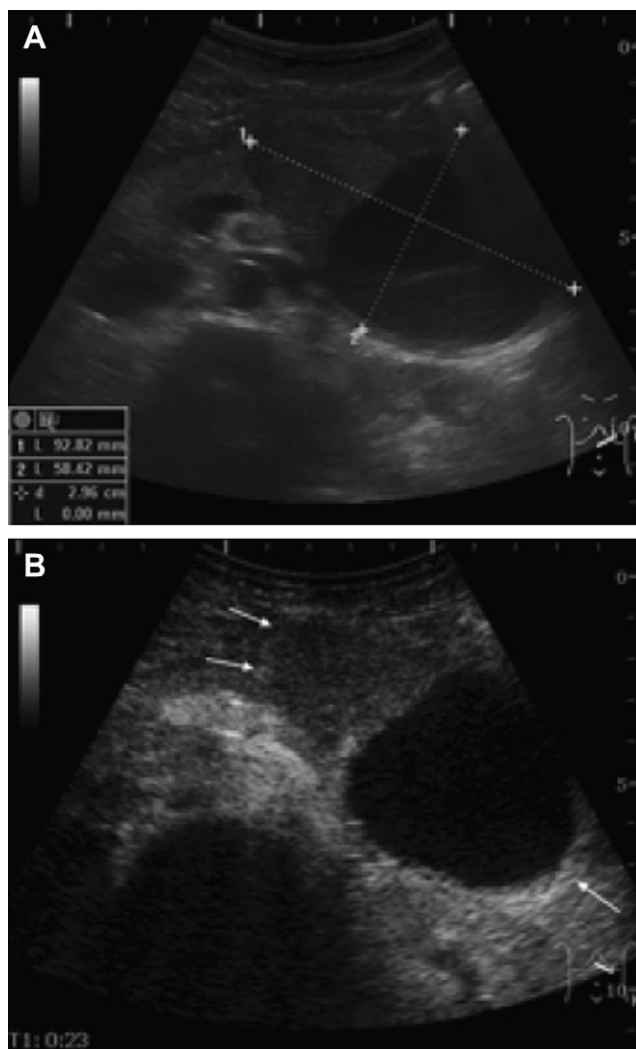


Fig. 1 (A) Ultrasound examination (transverse section) showing a well-defined mixed mass in the pancreatic body and tail. (B) Contrast-enhanced ultrasound (transverse section) showing homogeneous filling of the tumor in the arterial phase and the presence of peripheral rim enhancement (arrows).

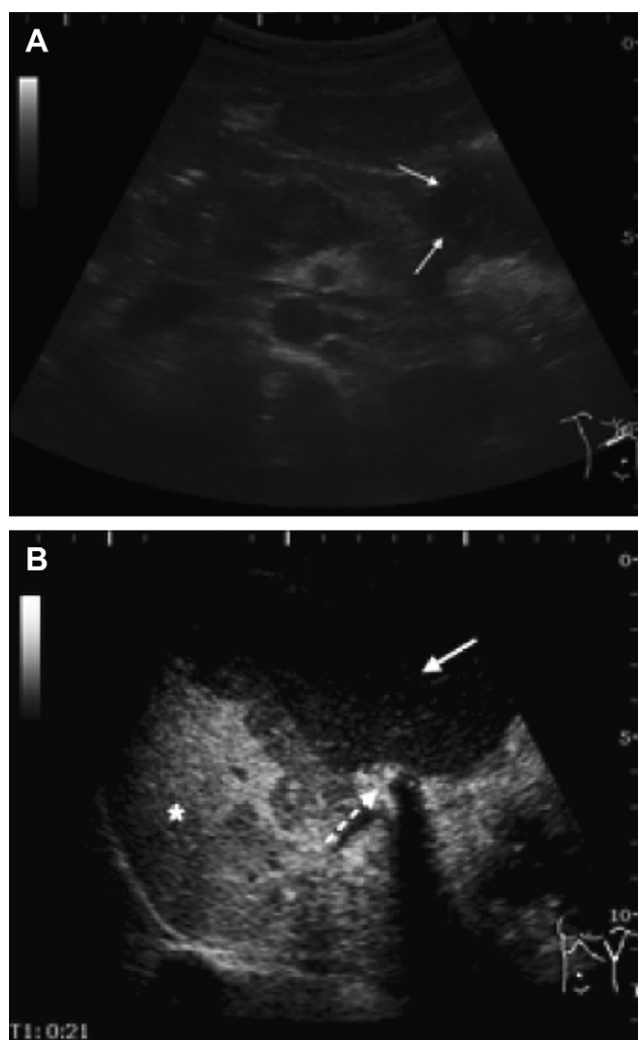


Fig. 2 (A) Postsurgical ultrasound examination (transverse section) revealed an inhomogeneous pancreatic area and the presence of a hematoma (arrows). (B) Postoperative contrast-enhanced ultrasound examination of the spleen (longitudinal section) showed normal spleen parenchyma (asterisk), splenic artery with contrast agent (pointed arrow), and infarction area (arrow).

diagnosis, SPT is usually a large tumor, with a reported mean diameter of 6.1–8.4 cm [4,7]. More frequently, this neoplasm is located in the body and tail of the pancreas (72%) [8]. The multicentric [9] or extrapancreatic forms [10,11] have rarely been described. Given the long doubling time, the prognosis of SPT is good even in the presence of metastasis [4], and therefore radical surgical treatment may offer a high survival rate.

CT and MRI are important imaging techniques that suggest the diagnosis of SPT. CT scan usually reveals an encapsulated tumor with possible areas of cystic degeneration, necrosis or hemorrhage [12]. Also, calcifications may be present. EUS-FNA has recently gained ground for evaluation of pancreatic tumors. The accuracy of EUS-FNA for diagnosis of SPT is superior to that of CT and MRI [13]. Also, it allows tumor biopsy, which permits immunohistochemical examination to differentiate SPT from other pancreatic

tumors. Contrast-enhanced harmonics endoscopic ultrasonography permits a more accurate description of tumoral vascular pattern [14].

US examination is frequently the first radiological technique in the management of these patients. The US aspects are not specific, but the clinical context and the evidence of a well-defined tumor may suggest SPT.

With CEUS, SPT has a homogeneous filling in the arterial phase and incomplete washout in the venous phase, due to the persistence of the contrast agent in the tumoral microcirculation. This specific behavior allows differential diagnosis from the high malignancy tumor, which remains more hypoechogenic during the arterial phase [15]. Frequently, the tumor is well-defined by a peripheral rim enhancement in the early dynamic phase. This corresponds to the tumor pseudocapsule, which results from the compression of the pancreatic tissue [16].

US examination may be useful in postsurgical follow-up to evaluate the local postoperative complications. In our case, US examination and CEUS revealed the presence of a postsurgical hematoma and spleen infarction. CEUS could be an important investigative technique to rule out active hemorrhage, as demonstrated by a lack of contrast agent inside the hematoma [17]. In addition to hemodynamic stability, these findings were decisive for a conservative approach in our case. Given the typical echographic aspect, US examination is accurate for the diagnosis of spleen infarction, whereas CEUS is more accurate for description of the infarction area [18].

Due to low malignancy potential, SPT is elective for radical surgical treatment, which provides excellent long-term survival. Distal pancreatectomy, with or without splenectomy, may be performed for tumors of the body or tail of the pancreas, and pancreaticoduodenectomy for tumors of the pancreatic head.

In conclusion, SPT is an uncommon pancreatic neoplasm, which may benefit from an excellent prognosis if diagnosed and treated surgically. US examination is often the first radiological technique and provides the first evaluation of pancreatic tumors. The combination of multiple US procedures (two-dimensional, Doppler and CEUS) improves the diagnostic accuracy and allows accessible follow-up in the postsurgical setting, to detect possible complications.

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